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Case Report

Broncho-biliary fistula secondary to biliary obstruction and lung abscess in a patient with pancreatic neuro-endocrine tumor



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Abstract We present a case report of broncho-biliary fistula that developed due to the blockage of biliary stent placed during the management of pancreatic neuroendocrine tumor (pNET); diagnosed on high clinical suspicion, percutaneous cholangiogram and contrast enhanced computed tomography (CECT); and successfully treated with percutaneous transhepatic biliary drainage (PTBD).

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Introduction

Broncho-biliary fistula (BBF) is defined as the passage of bile in the bronchi and the presence of bile in the sputum (biliptysis). BBF usually occurs either in the congenital form [1] or following thoraco-abdominal insult. BBF is a rare clinical presentation which was first reported in the year 1850 [2]. BBF has been commonly seen in hepatic malignancy [3,4]. Other causes of BBF include lung abscess [5], sub-phrenic abscess [6], postoperative stenosis (including biliary/gastric or pancreatic surgery [7]), amebic abscess [7,8], echinococcosis

[9,10], pyogenic liver abscess [11] and traumatic/surgical injury [12–14] to hepatobiliary system resulting in the obstruction of bile flow. BBF has been reported as post-surgical complication in many malignancies like cholangiocarcinoma [4], gall bladder carcinoma [13] and hepatic metastases [14] due to colorectal carcinoma [4], but to the best of our knowledge this complication has never been reported in clinical or therapeutic course of neuroendocrine tumor of pancreas.

Case report

A 37 year old female was diagnosed as a case of metastatic pNET when she presented with a severe backache in August 2012. She had a past history of lateral pancreaticojejunostomy for chronic pancreatitis in March 2012.

At the time of presentation, positron emission tomography-computed tomography (PET-CT) scan revealed an enhanced lesion in the region of the head and uncinate process

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of the pancreas with a few enlarged lymph nodes (LNs) along with multiple skeletal metastases. There was dilatation of middle and proximal common bile duct (CBD) with upstream bilobar intrahepatic biliary radical dilatation (IHBRD). She underwent endoscopic retrograde cholangio-pancreatography (ERCP) guided pancreatico-biliary stenting for obstructive jaundice. Fine needle aspiration cytology (FNAC) from peripancreatic LNs in September 2012 showed neuroendocrine tumor cells that were positive for synaptophysin and chromogranin-A. Ki 67 index was 4–5%, indicating a low grade tumor.

She received palliative radiotherapy and zoledronic acid for painful skeletal metastases. Patient received everolimus and octreotide therapy for six months. After progression on above therapy patient received peptide receptor radiotherapy (PRRT), four sessions till October 2014.

In January 2015 she presented with the complaints of fever and cough for one month. Radiological evaluation revealed left sided loculated empyema with concomitant left sided lung abscess (Fig. 1). Intercostal drainage tube was placed for the drainage of loculated empyema (Fig. 2) along with single aspiration of lung abscess. Microbial culture of lung abscess showed the growth of *Escherichia coli* (*E. coli*). Antibiotics were started accordingly. Although fever subsided cough aggravated with the production of green colored sputum (more

of serous in consistency) and patient was unable to lie down due to aggravation of cough. High resolution computed tomography (HRCT) of chest showed a thick walled cavity lesion in the antero-lateral-basal segment of the lower left lobe with loss of fat planes with the diaphragm.

Contrast enhanced computed tomography (CECT) of the abdomen (Fig. 3) revealed dilated intrahepatic biliary radicles with heterogeneous enhancement in segment II of the liver. There was a loss of fat planes between the involved hepatic segment, adjacent left dome of diaphragm and the basal segment of lung suggestive of possible contiguity.

Percutaneous cholangiogram (Fig. 4) showed biliary ductal dilatation and subtle contrast leak from left hepatic duct with pooling in adjoining area (left lung base) along with blockage of previously placed biliary stent.

In view of the above findings and increasing discharge of green colored sputum of 400 ml/day, biliptysis due to broncho-biliary fistula was suspected. Percutaneous transhepatic cholangiogram was done. Post percutaneous transhepatic biliary dilatation (PTBD), patient showed a marked improvement in symptoms. Biliptysis stopped immediately following PTBD procedure. At one week follow up, ERCP cholangiogram was done which did not show broncho-biliary fistula, indicating spontaneous sealing of fistula. CBD stent was replaced. PTBD tube was removed after 48 h of ERCP placed

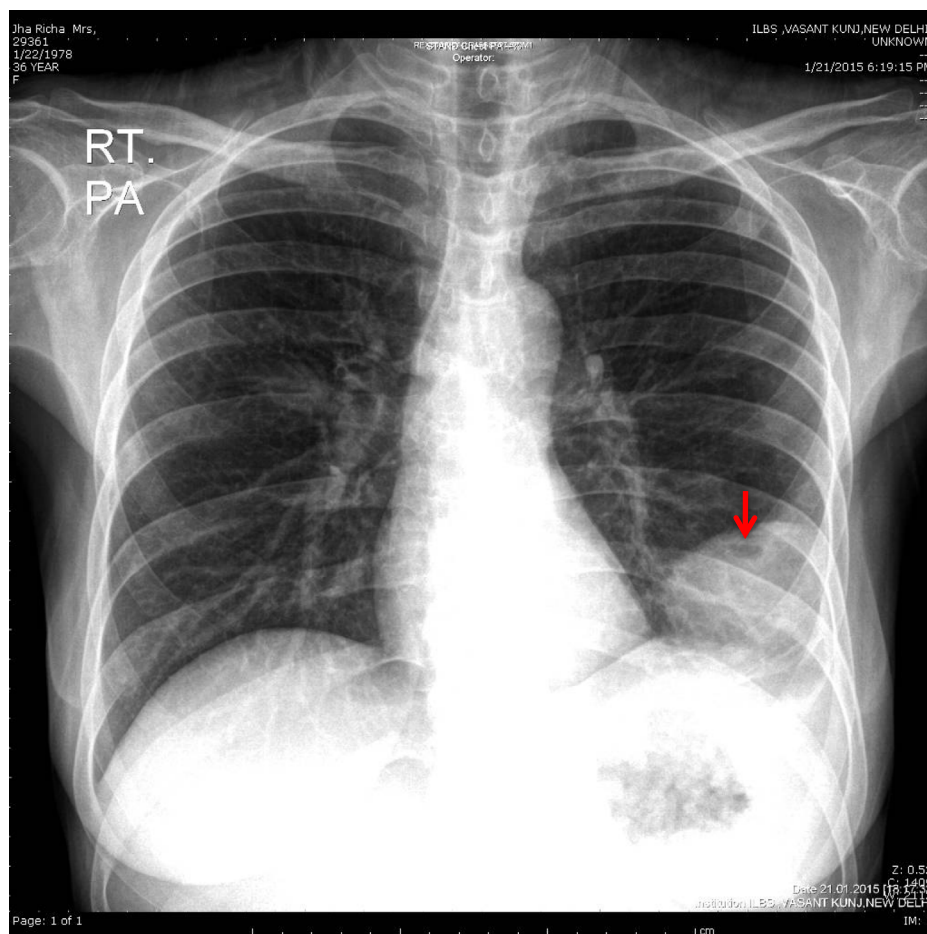


Figure 1 X-ray chest showing pleural based well defined, homogenous opacity with air fluid level within (arrow) indicative of loculated empyema with underlying lung abscess.

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